

Unusual case of disappearing hepatic hydatid cyst: COVID-19 times

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SUMMARY

The larvae of the tapeworm *Echinococcus granulosus* cause echinococcosis or hydatid disease. Liver is the most commonly involved organ accounting for 60%–80%. Rupture into the biliary radicle through a cystobiliary communication is the most anticipated complication seen in 5%–17%, presenting with obstructive jaundice and cholangitis. We present a young patient who had presented with cholangitis and a tender hepatomegaly. Contrast-enhanced CT (CECT) had revealed a large hepatic hydatid cyst with multiple daughter cysts and significant dilatation of intrahepatic/extrahepatic biliary system. He had undergone an emergency Endoscopic Retrograde Cholangiopancreatography (ERCP) with extraction of the membranes and stenting of the common bile duct. A few cycles of albendazole were given to sterilise before elective laparoscopic surgery. However, a follow-up CECT showed complete resolution of the hepatic hydatid with calcification. In conclusion, medical treatment with a few cycles of albendazole and ERCP with stenting could be a good treatment option.

BACKGROUND

The reason we wrote this case report is that it is a common problem in the Indian subcontinent. Complicated hydatid disease of the liver is always treated with surgery and closing the cystobiliary communication. Here, we show an emergency drainage procedure of the common bile duct (CBD) due to cholangitis and pharmacotherapy alone could be curative. This is relevant in very sick patients.

CASE PRESENTATION

A young patient had presented with a short history of right upper abdominal pain, high-grade fever with chills/rigor and deep jaundice. He also gave

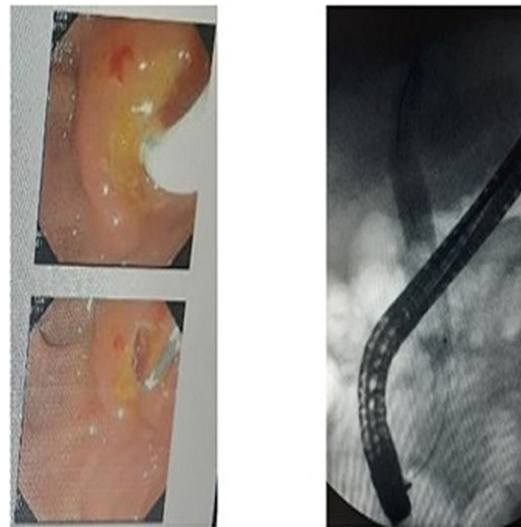


Figure 2 Endoscopic Retrograde Cholangiopancreatography (ERCP) showing hydatid membrane and a 7F double pig tail stent in the common bile duct.

a history of high coloured urine but not associated with pale stools or bleeding tendency. On further questioning, he had non-specific dyspeptic symptoms lasting for more than a year. Being from a village background, the joint family depended on agriculture for their livelihood. They also had cows and goats at home many years back. He would regularly take the goats for grazing in the neighbouring forest along with his dog. Examination revealed a toxic looking patient with fever of 38.3°C, tachycardia at 112/min, blood pressure was 126/84 mm Hg, a respiratory rate of 18/min. He had significant icterus and a tender hepatomegaly with localised signs of peritonism in the right upper quadrant. Per rectal examination was normal. The systemic examination was normal.

INVESTIGATIONS

Our routine blood workup had shown neutrophilic leucocytosis, an obstructive liver function test with a total bilirubin of 11 mg/dL and direct of 8.04 mg/dL. Ultrasonography (USG) of the abdomen had shown a large cyst in the liver with intrahepatic biliary radicle dilatation and no gallstones. With a normal serum creatinine, an urgent contrast-enhanced CT (CECT) scan of the abdomen had shown a large hepatic hydatid cyst (HHC) measuring 12×8 cm enclosing multiple daughter cysts involving segment IV, VIII, caudate lobe with dilatation of the intrahepatic and extrahepatic biliary system ([figure 1](#)). The

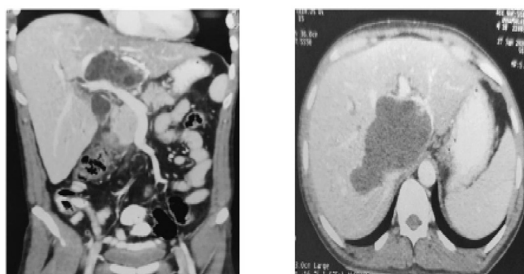


Figure 1 CECT showing a large hepatic hydatid cyst with multiple daughter cysts involving segments IV, VIII and caudate lobe along with dilatation of the intrahepatic and extrahepatic biliary system. CECT, contrast-enhanced CT.



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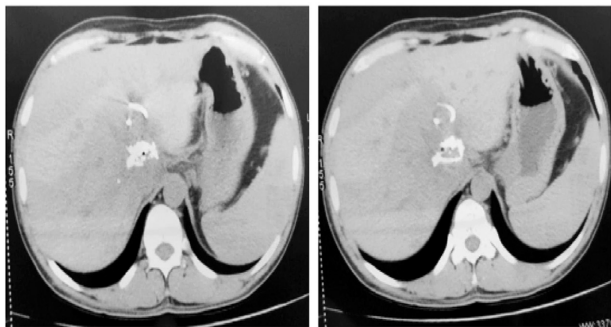


Figure 3 Postpulse therapy CECT demonstrating complete resolution of the hepatic hydatid cyst with calcification. CECT, contrast-enhanced CT.

patient had undergone a diagnostic and therapeutic Endoscopic Retrograde Cholangiopancreatography (ERCP) with endoscopic sphincterotomy and extraction of hydatid elements along with drainage of purulent bile from the CBD using a 7F double pig tail stent (figure 2). Bile aspiration during ERCP had grown *Escherichia coli* sensitive to piperacillin/tazobactam.

DIFFERENTIAL DIAGNOSIS

Our working clinical diagnosis was amoebic liver abscess with secondary bacterial infection, we see this often in this part of India (3–5 cases a month). Clinically, we also suspected choledocholithiasis causing cholangitis and pyogenic liver abscess. However, radiology revealed a classical hepatic cyst with a cart-wheel pattern and dilated biliary system. So the final diagnosis was a complicated HHC with rupture into the biliary system.

Further, this was proved while performing an ERCP where hydatid elements were extracted using a rat toothed forceps and purulent bile was emanating from the CBD.

TREATMENT

The patient had undergone a therapeutic ERCP with endoscopic sphincterotomy and extraction of hydatid elements along with drainage of purulent bile using a 7F double pig tail stent (figure 2). Intravenous piperacillin/tazobactam antibiotic was administered for 7 days. Simultaneously pulse therapy of albendazole 400mg two times per day for 21 days was started for two cycles with 1 week rest in between. This would sterilise the HHC and prevent anaphylaxis during laparoscopic evacuation. However in the ensuing 2 months the patient came back once with cholangitis secondary to a blocked stent. A repeat ERCP with stent exchange was done under antibiotic cover. Unfortunately, SARS-CoV-2 pandemic broke out and all elective surgeries were postponed as per government guidelines. As a result, we had advised him to continue with pulse therapy of albendazole through teleconsultation.

OUTCOME AND FOLLOW-UP

When elective surgeries resumed, the patient was readmitted following a negative COVID-19 real time reverse-transcription PCR report and he was asymptomatic. A routine preoperative CECT was done, and to our surprise, there was complete resolution of the HHC with calcification (figure 3). The patient had fully recovered thus obviating the need for surgery! As a result, we removed the stent before discharge. On short follow-up at 3 and 6 months, he was doing well clinically and USG abdomen shows no recurrence. He is due for a 6 monthly review.

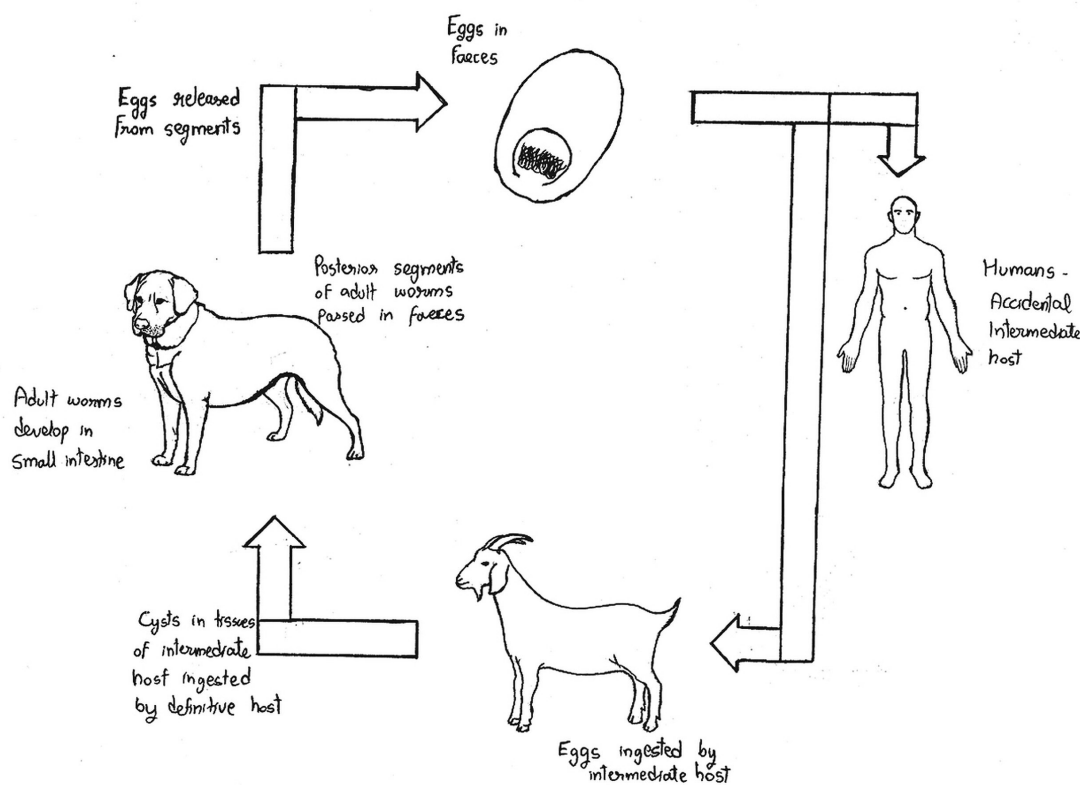


Figure illustrated by Shrenik Govindaraj

Figure 4 Life cycle of *Echinococcus granulosus*.

DISCUSSION

Echinococcus is a zoonotic disease and its prevalence is directly linked to contact with dogs and sheep. Human beings are intermediate or accidental host through ingestion of egg contaminated faeces (figure 4). The larvae is liberated in the duodenum where it invades the mucosa and reaches the liver through the portal circulation occupying the right lobe in 70%. This is due to the streamlining flow of portal blood anatomically. In the liver, it is transformed into a cyst. It differentiates into an ectocyst, a dense fibrous reaction by the host and an endocyst which has an outer laminated membrane and an inner germinal layer, which is the active part that gives rise to the brood capsules containing scolices. As the cyst grows, the intracystic pressure ranges between 80 and 120 cm of H₂O causing pressure necrosis of the biliary radicle producing a cystobiliary communication releasing the cyst content.¹ However, it can rupture into the bronchial tree, peritoneal cavity or produce pressure on adjacent organs. Uncomplicated cysts are very indolent in their growth causing vague abdominal pain due to the capsular distension. The diagnosis is based on serology and CECT, which shows the classical cartwheel appearance of the liver cyst. ERCP is indicated if we clinically suspect a biliary communication,¹ and it has a high success rate of cure.² Preoperative endoscopic sphincterotomy has shown to reduce the incidence of biliary fistulae and its associated morbidity.³ HHC can be managed by operative or nonoperative methods. Surgery remains the 'gold-standard' treatment. Surgery aims to inactivate the parasite, evacuate the cyst cavity, remove the germinal membrane and obliterate the residual cyst cavity. Non-operative methods include chemotherapy, percutaneous puncture aspiration injection reaspiration (PAIR).⁴ A couple of cycles of chemotherapy preoperatively sterilises the cyst and reduces intracystic pressure thereby preventing anaphylaxis. Benzimidazoles like mebendazole, albendazole are used to treat hydatid. They interfere with the absorption of glucose, depleting glycogen in the mitochondria and endoplasmic reticulum causing degenerative changes.⁴ A recent meta-analysis of 33 studies concluded surgical outcomes were better when benzimidazole were added.⁵ The pulse therapy varies from 4 to 6 week phases of albendazole at 10–15 mg/kg/day with a 1 or 2 week rest of 3–6 cycles.^{6–7} There are studies with albendazole used as a standalone agent for a prolonged period showing a good resolution in 73.7%.⁸ Percutaneous treatment of HHC was introduced in 1985 by Mueller *et al* as an alternative to surgery and medical therapy.⁹ Even though three decades have passed, controversies persist regarding its use as a primary treatment modality. There are a few studies favouring PAIR as an initial step in treating HHC with good results in terms of shorter hospital stay, complications, effectiveness, economical and disease recurrence.^{10–13} These results are very promising but have severe shortcomings as they are small series, retrospective or have a short follow-up. So in 2006 a Cochrane data systemic review assessed the merits and demerits of PAIR with sham/no intervention, surgery or medical treatment in uncomplicated HHC. The authors concluded that PAIR seems promising, but there is insufficient evidence to support or refute it as a primary treatment option and suggested further randomised clinical trials.¹⁴ WHO also recommended PAIR for management of uncomplicated HHCs in selected cases.^{14–15} But many authors have experienced problems regarding efficacy and safety with multivesicular cysts with high failure rates of up to 30%.^{11–16} Objectively, most studies including the WHO agree PAIR are contraindicated when cysts communicate with the biliary tree. Patients who received pulse therapy should be thoroughly reassessed of their

symptoms, blood profiles and a CECT scan repeated before considering elective surgery. Surgery should not be considered as a routine without reassessment. This is even more relevant in our case because of the prolonged course of pulse chemotherapy. Our case reiterates the fact that prolonged pulse therapy with albendazole and ERCP clearance of CBD with stenting could be an effective standalone treatment option in patients with complicated HHC with biliary rupture.

Patient's perspective

I was very satisfied with the entire treatment in the hospital and the final outcome. In a way I was happy, the corona pandemic helped me with the medical rather than a surgical treatment. I was shocked to understand from the doctors, how the livestock which I had regularly grazed more than a decade back brought about this disease. I am very grateful to the entire medical team who treated me. I equate them to near GOD status.

Learning points

- ▶ To consider a diagnosis of hydatid cyst in an endemic area.
- ▶ A patient with obstructive jaundice and cholangitis from an endemic area, we should consider a complicated hydatid cyst with rupture into the biliary system secondary to a cystobiliary communication.
- ▶ In an emergency scenario, an urgent therapeutic ERCP with clearing of common bile duct and stenting, combined with appropriate antibiotic is life-saving.
- ▶ A prolonged course of pharmacotherapy with albendazole can cure hydatid disease without resorting to surgery always.
- ▶ Patients on follow-up should always be systematically reassessed of their symptoms, blood profiles and a scan repeated before considering elective surgery.

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